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SHORT REPORT

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An unusual cause of stridor in childhood due to focal epileptic seizures

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Abstract Respiratory disorders with stridor are a frequent cause of admission for children in an emergency department. Laryngospasm, as an isolated symptom of epilepsy, is a rare phenomenon [1, 3, 5]. Other respiratory symptoms of epilepsy, rarely seen in childhood, might be apnoeic spells [2, 4]. We report on a child with laryngospasm due to focal epileptic seizures.

Keywords Child · Focal epileptic seizures · Laryngospasm · Stridor

A 19-month old boy was admitted twice in one week due to acute marked dyspnoea and sialorrhoea, which awakened the boy in the middle of the night. Clinical signs and symptoms were suspicious of acute laryngitis or epiglottitis and, at the first admission, the boy was intubated immediately. However, the laryngoscopy was normal, without signs of infection or swelling. The boy could be extubated within 6 h and was immediately free of any symptoms. Four days later he was readmitted with the same symptoms and was first treated with adrenaline inhalations without any success. Sudden cloni of the angle of the mouth and apnoea suggested seizure activity and after approximately three minutes of difficult mask ventilation benzodiazepine was given intravenously. The child recovered immediately from all symptoms. The stridor wasn't marked enough to think of an obstructive apnoea and the evolution after benzodiazepine enforced the hypotheses of seizure activity.

The EEG performed the day of the second admission showed a focal theta activity fronto-temporally and there was focal slowing in the fronto-temporal right-sided region. There were two episodes of 110 s duration with start of rhythmic theta activity in the right temporal area, spreading over the right hemispheres and finally to the left hemisphere (Fig. 1). Clinically, there was a short stop in crying after the beginning of epileptic activity, not accompanied by tonic-clonic or other obvious seizure activity. Level of consciousness during these episodes was uncertain. Magnetic resonance imaging (MRI) showed no structural abnormalities. In the personal history of the boy, there was a correction of Fallot-Tetralogie at the age of six months with an uncomplicated postoperative follow-up. Otherwise, this was a healthy boy, who achieved his developmental milestones. He is the first child of healthy parents and no neurological problems are known in the family.

The patient was treated with carbamazepine. He showed no further seizure activity in the follow-up of one year and EEG findings normalized.

Laryngospasm, as a main manifestation of epilepsy, is a rare phenomenon. Reviewing the literature (PubMed, Medline. Keywords: epilepsy; epileptic phenomenon; laryngospasm; child), only seven reported cases were found [1, 3, 5]. The most important differential diagnosis is infectious, allergic swelling or spasm of the larynx and/or trachea. Other causes of laryngospasm, such as hypocalcaemia or hypomagnesaemia, can easily be ruled out and are encountered infrequently.

Innervation of the larynx is by the Xth cranial nerve via the superior and inferior laryngeal nerve branches, however the pathophysiology of epileptic stimulation is not known [3]. For other respiratory symptoms, mainly apnoeic seizures, ictal records reveal epileptic activity in the same area as in our patient [2, 3]. The cortical motor representation of the larynx has been outlined by transcranial magnetic stimulation [6]. Laryngeal seizures are thought to originate from lower motor homunculus and insular area, which would be supported by our EEG-findings. However, this region receives afferent input from sensory, premotor and limbic areas leaving the actual site of epileptic origin a matter of speculation. As the MRI showed no structural abnormalities, the

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Fig. 1 Bioelectrical seizure onset: a 4- to5-Hz discharge pattern develops from right temporal electrodes (F8-T4) and spreads out into the right rolandic area (C4-P4). *Arrow* indicates the beginning of the epileptic discharge

congenital heart failure and its surgical treatment were not considered to be related to this epileptic problem.

In our patient, and in those reported earlier, the antiepileptic treatment showed a prompt response. This supports the assumption that laryngospasm has to be considered an epileptic phenomena.

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